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Case Report

Hematidrosis and Hemolacria: Report of Two Cases From Indonesia

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ABSTRACT

Hematidrosis and hemolacria are both extremely rare clinical entities with different etiologies. The authors herein report two cases of both hematidrosis and hemolacria in an 11-year-old girl and a 21-year-old man. While the etiology of these disorders are still unknown, there are a large number of associated disorders linked with hematidrosis and hemolacria mainly psychological distress, vicarious menstruation and rarely pathological conditions.

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Introduction

Hematidrosis is an extremely rare clinical phenomenon in which an individual sweats blood while hemolacria occurs when an individual sheds bloody tears.[1, 2] Manonukul and colleagues described this disorder as "hematofolliculohidrosis" encompassing sweat-like fluid and the blood coming out from follicular canals.[3] The etiology of this entity still remains a mystery although some theories have been proposed.[3-5]

This disorder has been mostly reported under acute fear, intense mental contemplation and excessive stress. Each episode is usually self-limiting, meanwhile physical and laboratory investigations do not lead to any abnormal results.[3, 6] We report two cases of hematidrosis and hemolacria which were treated successfully.

Case Presentation

Case 1

An 11-year-old Indonesian girl reported sudden onset of bleeding from both her eyes, left palm, the right nostril and left forehead (Figure 1). There was no trauma, wound, or skin defects. The bleeding stopped spontaneously according to the parents. However, one month later, the bleeding recurred from her right forehead, right cheek, left eye, left nostril, and the mouth (Figure 2). Again, the bleeding stopped immediately until it relapsed in a shorter interval manifested as bloody tears and sweats on her forehead and areas under the nostrils and philtrum. At most, the bleeding episodes happened three times in a day. While bleeding, she experienced sharp burning sensation on her face and both of her eyes which lasted for some time. These abnormal sensations even persisted after the bleeding episodes had ceased. Her parents gave her cold compress to reduce the unpleasant sensation. There were neither inflamed eyes or vision loss during the bleeding episodes nor other bleeding manifestations such as epistaxis or gum bleeding. She already had started menarche around three months ago; however, with irregular intervals and duration without any menorrhagia episodes.

The family history was negative for such presentation. History was remarkable for milk protein allergy. She did

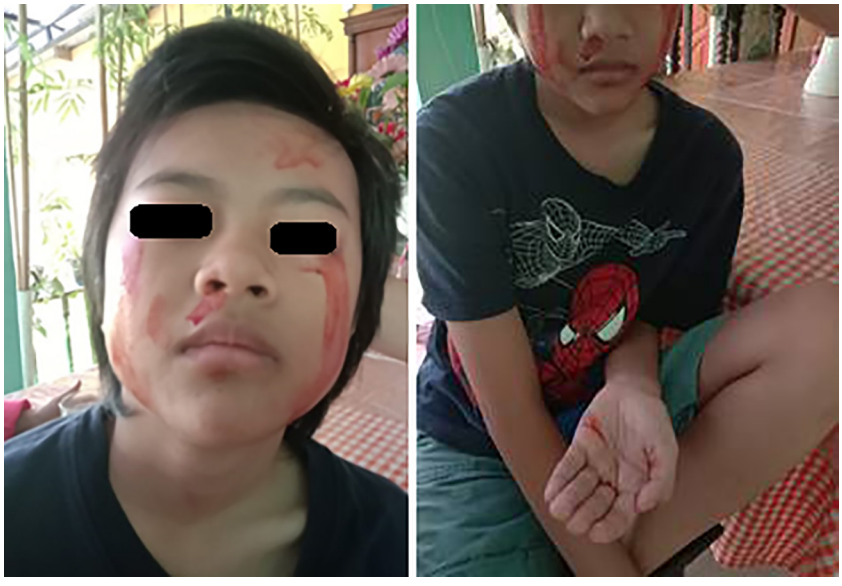

Figure 1: Bleeding under her eyes, right nostril and forehead (left) and from her left palm (right)

Figure 2: Bleeding from the right forehead and cheek (left) and from her left eye, left nostril, and around the mouth (right)

not have any history of treatment with anticoagulants, topical application of any medication or exposure to dyes. She was not also in any state of stress or psychiatric disorder. There was no problem at her school performance. Physical examination was otherwise unremarkable, and all laboratory and coagulation studies were within normal range.

Labetalol 10 mg twice daily for a month was prescribed for her. To this day, the symptoms had not relapsed.

Case 2

A 21-year-old male came to the outpatient clinic with repeated episodes of spontaneous bloody tears and sweat for one month. He had no history of epistaxis or gum bleeding. He did not have any prior significant medical history especially psycological stress or familial problems. Physical examination did not reveal any remarkable finding. Complete blood count and coagulation studies were within normal range. He was given multivitamin and reassurance for these conditions. Follow-up of the patient was negative for the recurrence.

Discussion

Hematidrosis is an eccrine sweat disorder presenting as one or several episodes of spontaneous, bloody sweating from non-traumatized skin.[7] Kluger and co-workers reviewed cases of hematidrosis from December 1996 to December 2016 and reported 25 cases. Most cases occured in women (84%) at the age of 8 to 30 years. Only eight (32%) cases were adults at the time of diagnosis with a median age of 13 years (range of 9-72 years old). Most of the cases (62%) were reported from Asia, while 25% were from Africa and only 4% were from Europe.[8]

This entity is tied to a religious belief as its cause. Hematidrosis is viewed as a stigma which formerly meant a spot, a sign, a wound, or a mark branded on a slave. From the time of Christ's crucification, this term got a special meaning as the reproduction of the wounds on the palms, soles and crown that Christ suffered on the cross.[2] As a proof that this entity had existed for centuries ago, Leonardo Da Vinci, as a prominent renaissance artist described a solider sweating blood before going to the battle. A famous Italian physician, Antonio Brassavola,

in the 16th century treated a nun who cried blood filled tears during her menstrual cycle.[6]

Historically, medicine is closely associated with superstitions, magic and religion and those beliefs are still held fervently until now, especially in Asia. Superstitions and stigmatization are tied together and are usually suggested as the underlying cause of hematidrosis.[9] The impact of the superstition and stigmatization is not to be undermined as widespread false beliefs and misdirected judgment may lead to delay in seeking treatment as parents or patients are ashamed and feared that their disease might get exposed.[10] This is especially true in hematidrosis, as the median time between onset of the symptoms and the first consultation was 6 months, with a range of a few days to 6 years.[8]

In the first case, it took almost more than one year for the parents to bring their child for a consultation, while in the second case, it took a month before seeking to the physician.

Pathogenesis of hematidrosis and hemolacria are poorly understood. The vasculitis in the dermal vessels and exacerbated sympathetic activation due to extreme stress and anxiety leading to peri glandular vessel constriction and subsequent expansion causes the blood seeps into the sweat ducts.[4] Uber and colleagues stated that there are multiple blood vessels arranged in a net-like pattern around the sweat glands. It is believed that under great stress, these vessels contract and then they dilate to the point of rupture. The blood at this point goes into the sweat glands, which pushes the blood to the surface and manifests as droplets of blood mixed with sweat.[11]

Another proposed theory is that there may be some defects in the dermis which cause stromal weakness. These defects will then communicate with vascular spaces in the dermis that will eventually dilate and enlarge as blood-filled spaces. Then, blood will leak out directly to the surface of the skin or via follicular canals. This will occur when there is enough positive pressure to force the blood out and later they will collapse, leaving no scar. This phenomenon acts like a balloon; waxes and wanes, and thus explains why these bleeding episodes are sometimes intermittent and self-limiting.[5]

Various conditions such as vicarious menstruation, thrombocytopenic purpura,[5] epileptic seizures, platelet factor 3 dysfunction,[6] psychogenic purpura,[12] psychiatric conditions such as oppositional defiant disorder[13] were also linked to the occurrence of hematidrosis. However, the exact mechanism is still unknown with no definite underlying condition.

Hematidrosis can be accompanied by otorrhea and otoerythrosis[14] as well as hemolacria.[2] Hemolacria has been reported secondary to giant papillary conjunctivitis, orbital varix, vicarious menstruation, conjunctival manipulation for follicle expression in trachoma, clinical treatment of conjunctiva with silver nitrate, epistaxis with retrograde blood stream into the conjunctiva through puncta lacrimalia and platelet factor 3 dysfunction.[2]

Face is the most commonly reported site of hematidrosis followed by the upper limbs, abdomen and pelvis, upper trunk, and the lower limbs.[8] The episodes are usually self-limiting and sometimes, the secreted fluid is more dilute and appears to be blood tinged, while others may have darker bright red secretions resembling frank blood.[11]

Diagnosis of hematidrosis can only be made if the following criteria are met: (i) recurrent, spontaneous, painless and self-limited oozing of bloody discharge confirmed by health professionals, (ii) existence of blood components in analysis of the discharge, and (iii) the site of bleeding is intact with no abrasion, telangiectasia or purpura and after wiping the area, there is no evidence of oozing. All of these criteria must be met in order to rule out organic bleeding disorders, self-inflicted bleeding, factitious disorder by proxy, and chromhidrosis (colored sweat)[6]. Biochemistry studies were not performed in our cases and only 2 criteria were met.

To date, there is no definitive management for hematidrosis, and it is important to investigate probable underlying conditions such as physical or psychological stress. Several drugs have been used such as propanolol,[13, 15-17] alprazolam,[5] lorazepam,[13] transdermal atropine,[6] and oxcarbazepine in case of hematidrosis with simultaenous epileptic seizures.[6] Vitamin C and hemostatic durgs were found not to be effective.[3] Non-pharmacological maneuvers such as relaxation, meditation, breathing exercises, psychotheraphy and behavioral interventions have been reported successful, albeit recurrence after stopping them has been reported.[8] In our cases, oral propanolol was successful in prevention from recurrence.

Clinicians should keep in mind that stigmatization and superstitions that tie hematidrosis to misleading belief in the public should be addressed and corrected. In this regard, counselling was reported to be more successful if combined with pharmacotherapy.[15]

Conclusion

Hematidrosis is an unknown disorder due to extremely low cases reported. It is of outmost importance to rule out underlying disorders and observe the spontaneity of the bleeding. Given the paucity of evidence available, it might be wise to look at the direction of possible underlying vasculitis as the probable cause. Parents and patients should be educated that the disease is most likely benign and transient in nature. All forms of stressors should be addressed and removed while onsidering all non-pharmacologic and pharmacologic measures.

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Ethical Approval

Ethical approval was not needed in this report; however, informed consent and permission to reproduce the data were obtained from both patients.

Conflict of Interest: None declared.

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